Cardiology in the Young

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Successful occlusion of a feeding artery with Amplatzer Piccolo Occluder in a patient diagnosed with Scimitar syndrome

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Brief Report

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Abstract

Scimitar syndrome is a congenital anomaly in which some or all of right pulmonary veins drain into inferior caval vein. It is associated with anomalous systemic arteries arising from descending aorta supplying to right lung. Transcatheter embolisation of this artery prevents complications. We present a 2.5-year-old girl in which anomalous artery was embolised using Amplatzer Piccolo™ Occluder, for first time.

Scimitar syndrome is a rare congenital anomaly in which some or all of the right pulmonary veins drain into inferior caval vein.[1] It is associated with other anomalies such as atrial septal defect, right lung and right pulmonary artery hypoplasia, anomalous systemic arteries arising from descending aorta supplying to the right lung, pulmonary venous obstruction, and dextroversion of the heart.[2] We present a case in which anomalous systemic artery was successfully embolised using Amplatzer Piccolo™ Occluder, for the first time in the literature.

Case report

A 2.5-year-old girl was admitted to our clinic with the complaint of recurrent respiratory tract infection. On physical examination, her weight was 12 kg and a grade I/VI systolic murmur was audible. Chest X-ray showed prominent pulmonary vascular markings at the right lower lobe. Echocardiography revealed anomalous pulmonary vein draining into inferior caval vein just below the diaphragm. CT showed hypoplasia of right lung and right pulmonary artery, dextroversion of the heart due to right lung hypoplasia, an anomalous arterial supply with three branches from descending aorta to a pulmonary sequestration of the right lower lobe and an anomalous drainage of right lower pulmonary vein to the inferior caval vein. With these findings, our patient was diagnosed as having Scimitar syndrome. Transcatheter device occlusion was planned. Catheterisation and angiography were performed by placing 5F sheaths in the femoral vein and artery. The anomalous vessel and its branch size measurements were obtained. Catheter angiography revealed return of left pulmonary veins and right upper pulmonary vein to the left atrium, right lower pulmonary vein to the right atrium at the junction of the inferior caval vein – right atrium, and sequestered tissue which was fed by non-tortuous feeding artery that separates from the abdominal aorta and extends to the right lower lobe (Fig 1), the venous return of the sequestered segment to the right atrium. Pulmonary artery pressure was 30/15 (23) mmHg, and Qp/Qs was 1.2. In angiographic measurements, the diameter of the feeding artery was 3.5 mm at its narrowest point, 4.5 mm at its widest point, and its length was 20 mm. We used a 4F multipurpose catheter to place a 0.014 inch guidewire into the feeding artery. Then, 5F delivery system was placed into the feeding artery via the guidewire. The Amplatzer Piccolo™ Occluder 5×6 mm device (with the largest disc diameter of 6.5 mm) was successfully implanted into the feeding artery via delivery system. Post-implantation angiography showed that the device was in a good position and did not protrude into aortic lumen, and there was no residual passage in the feeding artery through the device (Fig 2). The patient was discharged one day after the procedure. No complications were detected in the follow-up of the patient. She will be followed clinically for resolution of recurrent respiratory tract infections.

Discussion

Scimitar syndrome is associated with dextroversion of the heart, a Scimitar vein draining to the inferior caval vein, feeding artery arising from aorta. Coexistence of feeding artery with Scimitar syndrome is seen in 50 % of the cases.[3] Although the prevalence of Scimitar syndrome is low, it should definitely be considered in the differential diagnosis of recurrent lower respiratory tract infections. Transcatheter embolisation or surgical closure of the systemic arterial supply of the

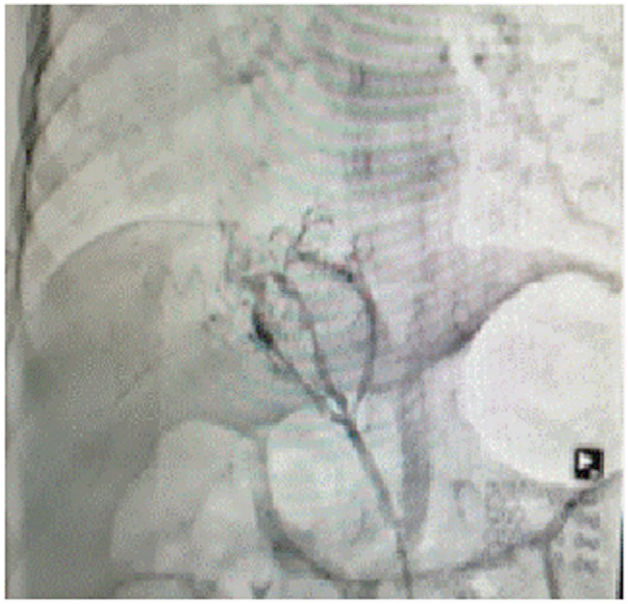

Figure 1. Pre-procedure angiographic view of the feeding artery and its branches.

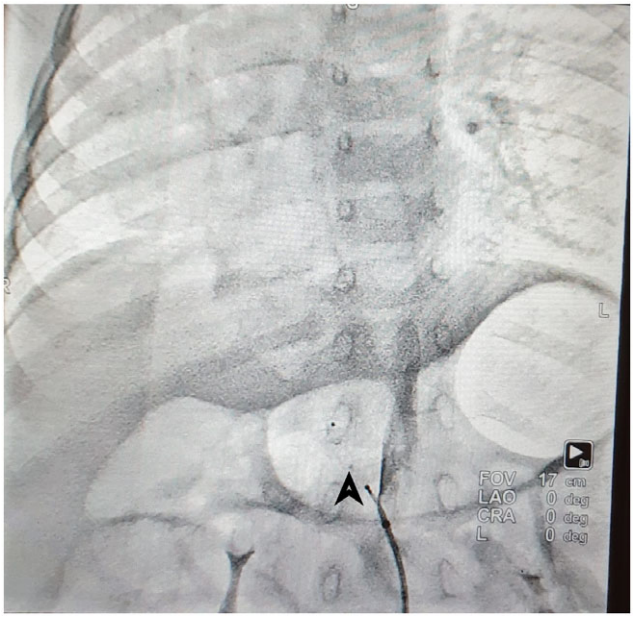

Figure 2. Post-procedure angiographic view of the feeding artery.

lungs prevents complications such as recurrent respiratory tract infections, pulmonary hypertension, and congestive heart failure. Traditionally, the treatment for pulmonary sequestration is surgery. However, surgery is associated with high morbidity and long hospital stay. Therefore, cases in which the feeding artery was closed with devices, such as coils, vascular plugs, or Amplatzer Duct Occluder and liquid embolisation such as N-butyl cyanoacrylate or Onyx, have been reported in the literature instead of surgery.[2,4,5] In 2002, Amplatzer Duct Occluder device was used for the first time by Crushell et all, to close the feeding artery.[6] In 2021, 2 cases diagnosed as pulmonary sequestration in which the feeding artery was embolised using Amplatzer Duct Occluder were reported by Zhang et al.[7]

In this case, the feeding artery was successfully closed with Amplatzer Piccolo Occluder for the first time in the literature. Embolisation with Piccolo Occluder is a safe and effective method and an alternative to other devices and to surgery for occlusion of anomalous vessels in the paediatric age group. The device can be placed in the feeding artery just as in the intraductal implantation of the Amplatzer Piccolo™ Occluder in ductus arteriosus of prematurity. We think that Amplatzer Piccolo™ Occluder is not suitable for closure of the feeding artery larger than 4 mm in width as the manufacturer's recommendations for use for closure of premature PDA (9-PDAP-05–06-L).

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Conflicts of interest. None

Ethical standards. The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guidelines on human experimentation (please name) and with the Helsinki Declaration of 1975, as revised in 2008. Informed consent was obtained from patient's family.

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